

CASE REPORT

Squamous odontogenic tumor: A case report and review of literature

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ABSTRACT

The squamous odontogenic tumor (SOT) is a rare, benign, locally infiltrative neoplasm of the jaws that appears to originate from the rests of Malassez, gingival surface epithelium or from remnants of the dental lamina. SOT was first described by Pullon *et al.* (1975). Since then there has been paucity in the number of reported cases, especially in the Indian subcontinent. The tumor is often asymptomatic, although it can present with symptoms of pain and tooth mobility. The characteristic radiographic appearance is that of a triangular-shaped unilocular radiolucency associated with the roots of erupted, vital teeth and has a predilection for the anterior maxilla and the posterior mandible. Histologically, the tumor is characterized by the formation of variably sized nests and cords of uniform, benign-appearing, squamous epithelium with occasional vacuolization and keratinization. We report a case of SOT occurring in a 58-year-old male in the anterior mandible with unusual localization and appearance.

Key words: Odontogenic tumor, rests of Malassez, squamous cells

INTRODUCTION

Squamous odontogenic tumor (SOT) is a lesion which had been recognized as an apparent entity for a number of years until 1975, when Pullon and coworkers published a series of six cases.^[1] SOT has been defined as a benign but locally infiltrative neoplasm consisting of islands of well-differentiated squamous epithelium in a fibrous stroma. The epithelial islands occasionally show foci of central cystic degeneration.^[2] SOT is a rare, benign but locally infiltrative epithelial tumor that supposedly develops from the remnants of dental lamina, or of the cell rests of Malassez (ROM), or gingival epithelium.^[3]

To date, fewer than 50 cases have been reported in the literature. The most common site of occurrence of the lesion in the mandible is the bicuspid-molar region and in the maxilla, incisor-cuspid area. Radiographically, SOT often exhibits a characteristic unilocular and triangular-shaped radiolucency of the alveolar bone, with the wide base of the radiolucency

localized between the diverging apices of the adjacent roots.^[4] Histologically, the tumor is characterized by multiple islands of squamous epithelium surrounded by a mature connective tissue stroma. Cystic degeneration or calcification may occasionally be observed in the epithelial islands.^[5] The usual treatment has been simple enucleation and recurrence has been rare.^[6] The purpose of this article is to report a new case and review the literature.

CASE REPORT

A 58-year-old male patient reported to our dental hospital with a complaint of swelling and mobility of teeth in the right anterior region of mandible from past 3 months. Onset occurred 3 months ago and it was asymptomatic. Medical history of patient was unremarkable. Clinical examination revealed a firm anterior swelling in right central and lateral incisor region of mandible. The mandibular right central incisor and canine were mobile. Radiograph [Figure 1] showed radiolucency with severe alveolar bone loss localized between right lateral and central incisor region of mandible, which was associated with markedly diverging roots. Based on the clinical and radiographic findings, the initial diagnosis of periodontitis was made and subjected for histologic examination. Histological examination [Figures 2–4] revealed a proliferation of mature stratified squamous epithelial islands in a dense fibrous connective stroma and the diagnosis of SOT was confirmed.

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Figure 1: Panoramic radiograph reveals triangular radiolucent area involving roots of mandibular right central and lateral incisor

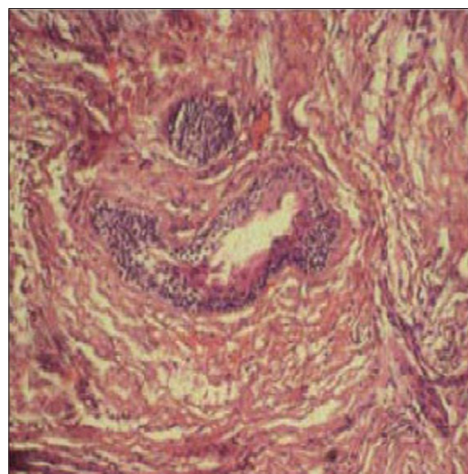


Figure 2: Photomicrograph revealing scattered islands of squamous epithelium in a dense fibrous connective tissue stroma (H and E, stain ×4 magnification)

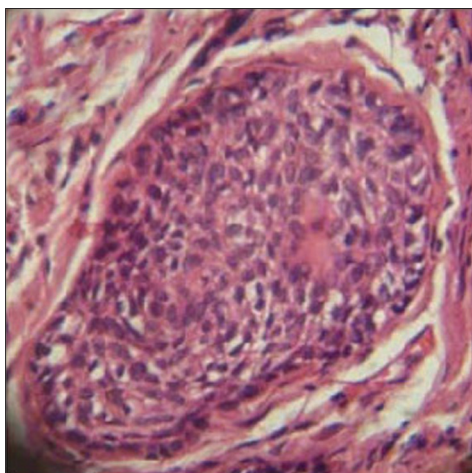


Figure 3: Photomicrograph revealing an island of benign squamous epithelium without presence of peripheral columnar cells, palisading nuclei or stellate reticulum (H and E, stain ×10 magnification)

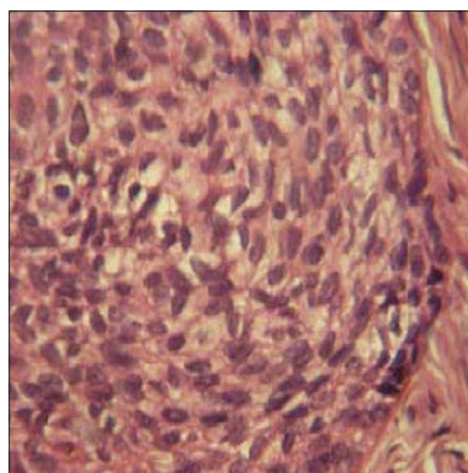


Figure 4: Photomicrograph of an epithelial island composed of uniform squamous epithelial cells (H and E, stain ×40 magnification)

DISCUSSION

After Pullon *et al.* had described an entity named SOT, a number of additional cases have been reported in the literature.^[3] To date, 44 cases of SOT have been reported [Table 1]. Due to the paucity of documented cases of SOT, the relative frequency among odontogenic tumors cannot be stated.^[2] SOT is known to occur in a wide age range, from first decade to eighth decade of life, with mean age of occurrence being 38.2 years. The gender ratio among 44 cases being 1:1.8 (F:M) showing slightly more predilection among males [Table 1].

Out of the 44 cases excluding our case, 20 have been located in posterior region of mandible. In maxilla, SOT occurred most commonly in canine-first premolar area.^[7] Our case is unique in that it is the first case reported in the Indian literature and second ever case to occur between the roots of central and lateral incisors in the anterior segment of the mandible. Hence,

the most common location for development of an SOT in the maxilla is anterior region and posterior in case of mandible with almost equal propensity to occur in both the jaws.^[7] SOTs occurring in maxilla were found to be more aggressive than in mandible.^[2,6] This was mainly due to the anatomy, porous and medullary nature of bone.^[8] Leider *et al.* reported a rare familial tendency for this neoplasm.^[9]

Most of the cases arise and develop in the periodontium of permanent dentition. One case was associated with the deciduous dentition and four cases were detected in edentulous areas.^[2-3,5]

Clinical presentation of SOTs represented a slow growing, intrabony lesion with very few clinical signs and symptoms. Most common being mobility of adjacent teeth, swelling of alveolar process, and mild/moderate pain in the affected areas. However, lesion may be asymptomatic and detected in routine intraoral radiographs.^[8]

Table 1: Summary of reported cases of SOT

Case	Reference	Age (years)	Sex	Site
1	Pullon <i>et al.</i> ^[3]	23	F	Multicentric
2	Pullon <i>et al.</i> ^[3]	11	M	Anterior maxilla
3	Pullon <i>et al.</i> ^[3]	19	M	Posterior mandible
4	Pullon <i>et al.</i> ^[3]	31	F	Anterior maxilla
5	Pullon <i>et al.</i> ^[3]	42	F	Anterior maxilla
6	Pullon <i>et al.</i> ^[3]	29	M	Posterior mandible
7	Doyle <i>et al.</i> ^[18]	26	M	Anterior maxilla
8	Doyle <i>et al.</i> ^[18]	65	M	Posterior maxilla
9	McNeill <i>et al.</i> ^[12]	26	F	Multicentric
10	Hopper <i>et al.</i> ^[19]	22	F	Multicentric
11	Carr <i>et al.</i> ^[10]	66	F	Posterior maxilla
12	Leventon <i>et al.</i> ^[20]	59	M	Posterior mandible
13	Kangvonkit <i>et al.</i> ^[21]	32	M	Anterior maxilla
14	Goldblatt <i>et al.</i> ^[22]	60	F	Posterior mandible
15	Goldblatt <i>et al.</i> ^[22]	29	M	Posterior mandible
16	Goldblatt <i>et al.</i> ^[22]	30	F	Posterior maxilla
17	Goldblatt <i>et al.</i> ^[22]	26	F	Posterior mandible
18	Goldblatt <i>et al.</i> ^[22]	67	F	Not stated
19	Cataldo <i>et al.</i> ^[23]	24	F	Anterior mandible
20	Swan and McDaniel ^[24]	32	M	Posterior maxilla
21	Norris <i>et al.</i> ^[25]	26	M	Post, max., bilat.
22	Kristensen <i>et al.</i> ^[26]	61	M	Ant. and post. maxilla
23	Monteil and Terestri ^[27]	51	F	Anterior mandible
24	Warnock <i>et al.</i> ^[28]	19	M	Anterior mandible
25	Mills <i>et al.</i> ^[29]	26	M	Multicentric
26	Tatemoto <i>et al.</i> ^[13]	41	M	Anterior mandible
27	Tatemoto <i>et al.</i> ^[13]	56	M	Anterior maxilla
28	Leider <i>et al.</i> ^[9]	29	M	Multicentric
29	Leider <i>et al.</i> ^[9]	25	M	Multicentric
30	Yaacob ^[30]	39	M	Anterior maxilla
31	Reichart and Philipsen ^[2]	56	M	Anterior mandible
32	Schwartz-Arad <i>et al.</i> ^[5]	8	M	Anterior mandible
33	Baden <i>et al.</i> ^[6]	46	M	Ant. and post.max.
34	Baden <i>et al.</i> ^[6]	39	M	Posterior mandible
35	Baden <i>et al.</i> ^[6]	74	M	Multicentric in maxilla
36	Saxby <i>et al.</i> ^[31]	59	M	Anterior mandible
37	Saxby <i>et al.</i> ^[31]	59	M	Anterior mandible
38	Saxby <i>et al.</i> ^[31]	59	M	Anterior mandible
39	Haghighat <i>et al.</i> ^[4]	43	M	Anterior maxilla
40	Barrios <i>et al.</i> ^[8]	11	M	Anterior maxilla
41	Ruhin <i>et al.</i> ^[15]	9	M	Anterior maxilla
42	Jwa Young <i>et al.</i> ^[16]	18	M	Posterior mandible
43	King kim <i>et al.</i> ^[7]	15	F	Posterior mandible
44	King kim <i>et al.</i> ^[7]	27	F	Posterior mandible

Anterior (ant.): Anterior to the distal surface of the canine tooth. Posterior (post): Posterior to the distal surface of the canine tooth

Radiograph of common central variant of SOT shows a well-defined unilocular, triangular radiolucency between the roots of adjacent teeth. Root resorption, radiopacities seen in other odontogenic neoplasms are seldom a feature of SOT.

Large and extensive SOTs may, however, show multilocular pattern. Root resorption was noticed in only one case.^[10] The peripheral variant may cause some saucerization of underlying bone. This was likely to be a pressure phenomenon rather than the result of true tumor infiltration.^[2]

Histologically, the lesion usually presents as islands of benign squamous epithelium in mature connective tissue stroma without the evidence of peripheral columnar cells, palisading nuclei, or stellate reticulum. The epithelial islands of SOT seem to resemble the squamous metaplasia seen in ameloblastoma; however, the lack of peripheral columnar cells and palisading nuclei establishes the differential diagnosis between these two tumors. Moreover, it must be distinguished from primary intraosseous squamous cell carcinoma, which is rare in young persons and presents dysplastic epithelial features.^[11] Keratin pearls, microcysts, and intraepithelial calcification are often present in SOTs. Circular areas of fibroblast and fibrous condensation/hyalinization can be seen around some of the epithelial islands, which suggests a connective tissue reaction to epithelial proliferation.^[7]

In one of the reported cases, electron microscopy showed epithelial cells with features similar to cells of stratum spinosum with numerous desmosomes, abundant glycogen granules, tonofilaments, and myelin bodies.^[12]

The pathogenesis of SOT has yet remained unclear. The epithelial odontogenic tumors are histologically related to remnants of odontogenic epithelium, which includes the dental lamina (cell rests of Serre), enamel organ, and the root sheath of Hertwig (cell ROM). Actively growing dental lamina is present within the jaws for a considerable time after birth. Because of the widespread presence of odontogenic epithelium, some tumors may arise from residues of these cells in bone or in soft tissues such as the gingiva. Regarding the pathogenesis of SOT, most researchers have pointed toward a periodontal ligament origin for the central variant of the lesion. Radiographic appearance of the lesion, occurring between the adjacent teeth in the tooth bearing area from the periodontal ligament space, and the pattern of bone destruction seeming to emanate relatively uniformly from the areas of tooth roots lend credence to the theory of origin from the periodontal ligament, viz., from the ROM. Microscopic findings, the epithelial islands which are intimately associated with the apices of the retained teeth, and tight adherence of tumor mass to the canine and ease of separation from the buccal soft tissue further add to the notion of origin from ROM. The ROM hitherto considered to be resting cells in the periodontal ligament have received much of attention recently. It is known that the ROM persists into adult life in all areas of the periodontal ligament but most abundantly in the area of the cervical half of the root where some have even proliferated and formed structures within the periodontal ligament similar to rests of Serre. The fact that some cases are reported in edentulous areas does not take away the possibility that they originated from ROM of teeth formerly present.^[4,9]

The heightened activity in nonperiodontal lesions can perhaps be attributed to certain genetic etiologies. The ameloblastin gene, for example, has been shown to express an important protein in epithelial-mesenchymal signaling during odontogenesis, and mutations of this gene have been found in SOT, Adenomatoid Odontogenic Tumor and Ameloblastomas. Further investigations need to be done to see if mutations in this gene have a vital role in tumorigenesis of SOT. In addition, the heparanase gene, which codes for an enzyme that is important in dissemination and cellular invasion, is prevalent in ameloblastomas. The production of heparanase was thought to contribute to its local invasiveness. Further studies with heparanase in other locally invasive lesions such as SOTs can perhaps be performed to establish additional genetic influences related to the aggressiveness of some of SOTs reported recently.^[7]

Carr *et al.* postulated a gingival origin for the SOTs and observed that the insular growth pattern of the tumor, the presence of squamous cells with clear cytoplasm, the parakeratosis, and the tendency to microcyst formation were related to gingival rests of Serres.^[10]

Proliferation of squamous epithelial islands in SOT has also been reported in submucosal regions, which might indicate that the lesion originates from the rests of Serres. However, it should be recognized that there are peripheral variants of SOT that are thought to arise from gingival surface epithelium or from remnants of the dental lamina (rests of Serres).^[10]

SOT-like proliferations (SOT-LP) have been reported in fewer than 15 cases, arising from the walls of the odontogenic cysts. These lesions were cystic by nature, although histologically were similar to SOT, they exhibited no tendency to evolve into neoplasms. SOT-LP exhibited varying clinical, radiological, and prognostic features than true SOTs. Biologically, SOTs are less aggressive than ameloblastomas but are more aggressive than SOT-LP in the cysts, except for those arising from the walls of odontogenic keratocysts.^[9]

Immunohistochemical studies conducted by Tatemoto *et al.* and Yamada *et al.* on SOT confirmed the proliferative activity of odontogenic epithelium indicated by heavy staining for keratin 13/16, and squamous differentiating cells in the center of tumor islands have shown a strong positive reaction for involucrin staining.^[13,14]

Treatment of SOT involves conservative local excision, curettage, enucleation, and scaling of adjacent teeth.^[2,8] Recurrences have been reported in only one case, most likely due to insufficient initial removal.^[8]

Ide *et al.* suggested that SOT could transform into a malignant disorder such as intraosseous squamous cell carcinoma.^[17] King Kim *et al.* suggested that SOT can decorticate the dense bone of mandible and may impact the therapeutic modalities

and diagnosis management of these presumed innocuous lesion.^[7]

SOT is a benign odontogenic neoplasm of distinctive histological features probably arising from ROM, although three decades have passed since the establishment of the lesion as a separate entity much remains to be learned about the SOT. The sporadic reports in the literature belie the propensity of the lesion to occur in the jaws. A probable reason could be the comparative few reports and the common mistaken diagnosis of an acanthomatous ameloblastoma.

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